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CASE REPORT

Anterior Stafne bone defect mimicking a residual cyst: a case report

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Stafne bone defects (SBDs) are asymptomatic lingual bone depressions of the lower jaw that are frequently caused by soft tissue inclusion. The common variant of SBDs exists at the third molar region of the mandible below the inferior dental canal and has been mostly diagnosed incidentally during routine radiographic examination. The anterior variant of a SBD (ASBD) is relatively uncommon and is located in the premolar region of the mandible. Sublingual salivary glands are thought to be responsible for ASBDs. However, other structures such as lymphoid or vascular tissues might be associated with ASBDs. In the present report, an ASBD which was mimicking a residual cyst was diagnosed with the aid of a three-dimensional CT scan. ASBDs might be confused with other odontogenic or non-odontogenic pathologies because of their location and lower occurrence rate. Advanced imaging modalities, especially CT scans, are useful to assess such lesions in order to avoid unnecessary surgery.
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Case report

A 62-year-old otherwise healthy woman was referred to our clinic for routine dental examination and prosthetic management. Panoramic radiograph of the patient revealed a well-defined radiolucent area at the right premolar region of the mandible below the root of the canine tooth (Figure 1). The overlying mucosa of the lesion was quite normal and there was not any sign of infection or fistula. The lesion was unilocular with a diameter of almost 2 cm, presenting radiopaque margins mainly in the upper limit. The location of the lesion was relatively far from the alveolar crest and it was thought to be a residual cyst as the premolar region of the patient was edentulous. However, the lesion was asymptomatic and no history of expansion was recorded. Axial, coronal and three-dimensional CT scans of the patient showed a lingual bony defect at the right premolar region (Figure 2a,b). The lesion was diagnosed as an anterior variant of a Stafne bone defect (ASBD). The patient was informed about the lesion and scheduled for follow-up.

Discussion

Stafne bone defects (SBDs) were first described by Stafne as usually unilateral, asymptomatic, well-defined radiolucent lingual bony defects located at the posterior region of the mandible below the inferior dental canal.[1] SBDs have also been called lingual bone defects, Stafne's bone cavity, static bone cyst and idiopathic bone cavity. This type of lesion has been known as the posterior variant of SBDs, with an occurrence rate of 0.1–0.48%.[2,3] In 1957 Richard and Ziskind[4] reported a lingual bone defect that was located anterior to the molar region. Since then, almost 40 cases of anterior mandibular salivary gland defect have been reported in English literature.[5] ASBDs are more prevalent among males and the incidence of ASBDs is quite rare (less than 0.009%)[6] compared with the posterior variant of SBDs. ASBDs are usually located at the premolar and

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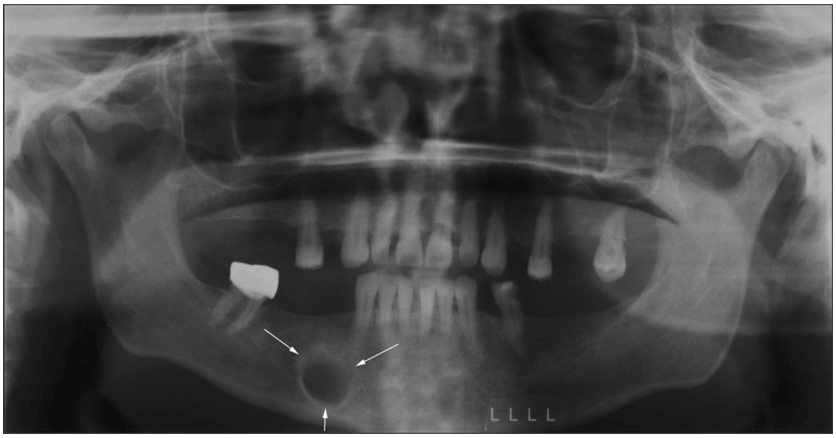
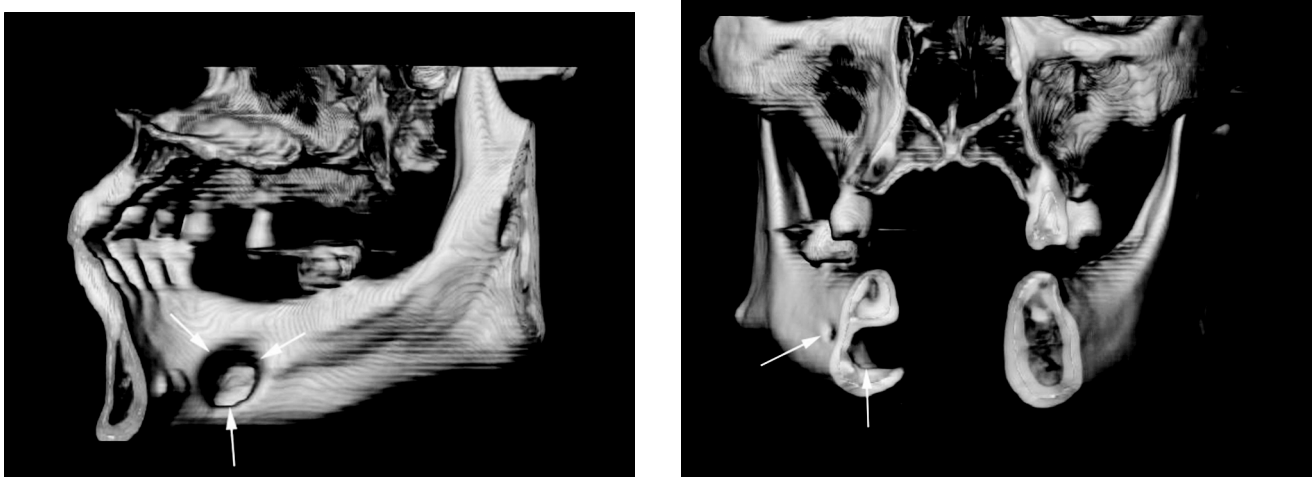


Figure 1 Panoramic view of the lesion in the anterior mandible

canine regions of the mandible near the first molar teeth and are prone to being confused with other jaw lesions. However, it has been reported that almost 17% of reported cases were seen in edentulous regions.[7] Thus, the initial diagnosis of ASBDs in edentulous patients might be a residual cyst, as in the present case.

The cause of the lingual bony defects is controversial. Stafne suggested that the occurrence of lingual cavities is developmental as the defect was occupied by cartilaginous tissue due to bone deposition deficiency;[1] however, some authors have suggested that pressure of the glandular tissue on the lingual cortex of the mandible causes a lingual bony depression.[8] According to this widely accepted concept, the submandibular salivary gland is responsible for the posterior variant of SBDs whereas the sublingual gland causes ASBDs.[7] Thus, many investigators found glandular tissue within defects either during surgical exploration or on MRI.[9] Additionally, Shibata et al[10] showed healing of the cavity after removal of the glandular tissue inside the lesion. Rare case reports have also been published regarding the muscular, lymphatic or vascular tissues related to the cavity.[11,12]

Lingual bone defects, mostly posterior variants, are usually diagnosed *via* conventional plain films since radiographic features of the aforementioned defects are frequently unique. However, in some cases, advanced diagnostic techniques such as sialography, CT, cone beam tomography and MRI might be needed to reach a definitive diagnosis. It has been reported that CT is a complementary diagnostic procedure for SBDs since other jaw pathologies could be distinguished with this method.[9] Segev et al[13] stated that diagnosis of SBD with CT is easier than with MRI, but they also mentioned that MRI should be considered to identify the content of the cavity. MRI is suggested for definitive diagnosis of SBDs and does not require exposure to ionizing radiation. On CT images, larger



a **b**

Figure 2 (a) Three-dimensional CT view of the lingual bone defect at the premolar region of the mandible. (b) Three-dimensional CT view of the defect located in front of the mental foramen

cavities of SBDs were perceived as being smaller than they actually were and the contents of cavities were uncertain.[14] MRI also has some disadvantages such as cost, discomfort to the patient and possible image distortion. Advanced imaging techniques such as CT and MRI are more useful for ASBDs since the differential diagnosis of anterior lingual bony defects might be more doubtful than that of posterior ones. In the present report, CT was preferred to identify whether the lesion was another jaw pathology or ASBD regardless of the content of the cavity. CT might be considered as the primary option to diagnose asymptomatic lingual bone cavities unless the content of the lesion needs to be investigated.

Sialography is also a diagnostic technique that can be used to determine whether glandular tissue exists in the cavity. However, this procedure is invasive and uncomfortable for patients.[15] In addition, sialographic evaluation of ASBDs is relatively hard to perform owing to multiple ducts in the sublingual gland.

ASBDs are a rare entity and they might be misdiagnosed as other pathologies such as sialadenosis, traumatic bone cyst, bone marrow defects, giant cell granuloma[5] or residual cyst.[7] The term residual cyst refers to an inflammatory odontogenic lesion that remains after removal of the related tooth.[16] However, growth of a residual cyst might also be caused by chronic low-grade inflammation over the course of several years that makes it asymptomatic. In addition, well-defined sclerotic bone margins are evident for cystic lesions, whereas radiographic borders of other pathologies such as giant cell granuloma or traumatic bone cyst would be more undefined. Although the present case of ASBD might have been misdiagnosed as a residual cyst, owing to its radiological appearance and location in the edentulous region of the mandible at the apical region of the previously extracted premolars, it might also have been confused with any other odontogenic or non-odontogenic lesioned.

In conclusion, advanced non-invasive radiological assessment with CT scans is usually sufficient to achieve a final diagnosis of ASBD and to avoid surgical interventions, which would be an unnecessary option in the management of ASBD except for symptomatic and/or concomitant other pathologies.

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